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Extrapleural Hydatid Disease of Chest: A Case of Recurrent Hydatid Disease

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Abstract

Hydatid disease is a parasitic infestation by a tapeworm of the genus Echinococcus. We present the case of 34 year old female who complained of chest pain and had a past history of hydatid cyst resection four times in last 15 years. She was found to have extrapleural hyatid cysts of chest that were removed via thoracotomy. The patient fully recovered and experienced an uneventful follow-up.

Introduction

Hydatid disease is a worldwide zoonosis produced by the larval stage of the Echinococcus tapeworm. Of the 4 known species of Echinococcus, 3 are of medical importance in humans. These are Echinococcus granulosus, causing cystic echinococcosis (CE); Echinococcus multilocularis, causing alveolar echinococcosis (AE); and Echinococcus vogeli. E granulosus is the most common of the three. E multilocularis is rare but is the most virulent, and E vogeli is the rarest. The two main types of hydatid disease are caused by E granulosus and E multilocularis. The former is commonly seen in the great grazing regions of the world — particularly the Mediterranean region, Africa, South America, the Middle East, Australia, and New Zealand — and is the most frequently encountered type of hydatid disease in humans. Theoretically, echinococcosis can involve any organ. The liver is the most common organ involved, followed by the lungs. These 2 organs account for 90% of cases of echinococcosis.[1] Clinical presentation is often non-specific and may be asymptomatic. Approximately, 60% have right hypochondrial pain and 15% become jaundiced. Other features include skin rashes, pruritus and allergic reactions.

Diagnosis can be made by Complete Blood Count (CBC) which will detect eosinophilia in 30% of patients, plain abdominal x-ray which may show calcification in cyst wall while the cyst can also be imaged with

ultrasound or CT. It can be confirmed by indirect haemagglutinin assay. Aspiration should not be performed if hydatid disease is suspected as it is associated with risk of dissemination of infection or anaphylaxis.[2,3]

Management in the form of pharmacological treatment is not curative but it is used as an adjunct to surgery to kill spilled scolices. The drugs of choice are albendazole, mebendazole and praziquantel. If surgery is required a laparotomy is performed to exclude other cysts.

Operative mortality is less than 2%. Complications include subphrenic abscess and prolonged cyst drainage.

Case Report

A 34 years old female presented with chest pain, cough and low grade fever. At the time of presentation she also had abdominal pain that had history of several years. She had history of resection of hydatid cysts from the abdomen 4 times in last 15 years. After resection of hydatid cyst for the fourth time she developed respiratory symptoms that turned out to be hydatid cysts in thorax and was referred to our center for further management. On examination she was vitally stable. CT abdomen was done that revealed widespread disease involving liver, kidney, spleen and pelvic structure. Therefore diagnosis of Hydatid disease was made and decision of thoracotomy and removal of hydatid cyst was taken.

Patient was placed in left lateral decubitus position and right posterolateral thoracotomy via 5th intercoastal space was performed. Extrapleural hydatid cysts were located in costophrenic angle, posterior to diaphragm. More than 800 cysts were removed. Cysts covered with thick exocysts were partially removed. Rest of the space was washed with hypertonic saline. Lung and pleura were normal. Extrapleural catheter was placed for pain control. Chest was closed in layers. Specimen of hydatid cysts were sent for culture and sensitivity.

Culture and Sensitivity revealed no microorganism, there were moderate pus cells seen. Significant numbers of β-hemolytic streptococcus group G were appreciated and treated accordingly post-operatively. Post-operative chest X-ray revealed right pleural effusion along with right chest basal atelectatic changes. Some subcutaneous emphysema was noted along the right lateral chest wall. Left lung was clear. Follow-up chest X-ray showed resolution of atelectatic changes on right side.

Marcaine was used as post-operative analgesia. Patient was sent to physiotherapy services for strengthening chest muscles. Post-perative course was uneventful.

Discussion

The diagnosis of hydatid disease in endemic areas is not very difficult to suspect. But the atypical sites involved by the disease may at times place hydatid disease down in the differentials as evidenced by case series published by Abu-eshy et al[4]. Though lung is second most common site involved[5] but extra-pulmonary hydatid disease is very rare that have been only scarcely reported in the literature[6]. Since the diagnosis of abdominal hydatid disease had already been established in our patient, it was actually clinical features of chest that made the case worth presenting. Chest radiography is considered to be most valuable first line of imaging study as reported by Dogan et al.[7] Since our patient was having recurrent abdominal hydatid disease therefore we opted for CT scan as imaging modality to determine the extent of abdominal and/or chest disease. Furthermore CT scan also helps to define the anatomy of disease. Since thoracotomy offers adequate simultaneous access to both the chest and hepatic lesions with acceptable morbidity and mortality, capitonnage provides no advantage in operations for pulmonary hydatid cysts as reported by Turna et al.[8] Therefore we performed standard thoracotomy in our patient. To the best of our knowledge there is no case of hydatid disease in costophrenic angle posterior to diaphragm reported yet. Though operative morbidity and mortality in hydatid disease is 0-13% and 0-5% respectively[9] and more clinical emphasis is on medical management of disease with Albendazole[10]. But surgical approach combined with chemotherapy in recurrent hydatid disease especially involving more than one organ system has been the cornerstone of management though other effective protocols of management combining chemotherapy, percutaneous drainage and surgery need to be design. We presented the case of extrapulmonary hydatid disease in an unusual location and with recurrence that was indeed a medical and surgical challenge to manage.

Conclusion

Hydatid Disease is prevalent in many parts of the world. Though highly variable in presentation it typically involves the liver but can involve virtually any organ system. Both chemotherapy and surgical options are available for management of hydatid cysts disease. Albendazole is active against Echinococcus. Invasive

options include percutaneous drainage along with scolicidal agents or more invasive but safer method is the surgical resection of cysts to avoid the rupture and consequent anaphylaxis.

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